CASE REPORTS

Refer to: Burbige EJ: Crohn's disease of the stomach. West J Med 126:397-400, May 1977

Crohn's Disease of the Stomach

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ALTHOUGH CASES had been described previously,[1] it was Crohn, Ginzburg and Oppenheimer who, in 1932, first recognized nonspecific granulomatous inflammation of the ileum as a separate disease entity.[2] They described 14 patients with the disease limited to the terminal ileum and hence the name regional ileitis was coined. However, since that time it has been shown that this nonspecific granulomatous process may involve many different sites in the alimentary tract from the mouth[3] to the anus.[4] In the last decade there has been an increased recognition of gastroduodenal involvement by Crohn's disease.[5-15]

Recently a patient was seen who initially was thought to have gastric carcinoma. The subsequent finding that she had a granulomatous involvement of the stomach prompted this review of the literature and consideration of some of the clinical features of the disease and methods of treating this disorder.

Report of a Case

A 42-year-old white woman, a hospital employee, had been in good health until March 1972. At that time excisional breast biopsies were done for fibrocystic disease. Shortly after the procedure she began to notice episodes of nausea, vomiting, anorexia and weight loss. By the time she presented to the outpatient department in December 1972, she had lost 30 pounds and complained of almost continuous epigastric gnawing pain. This pain was somewhat relieved by taking antacids. However, the episodes of nausea and vomiting had increased in frequency to the point that she was unable to eat without vomiting.

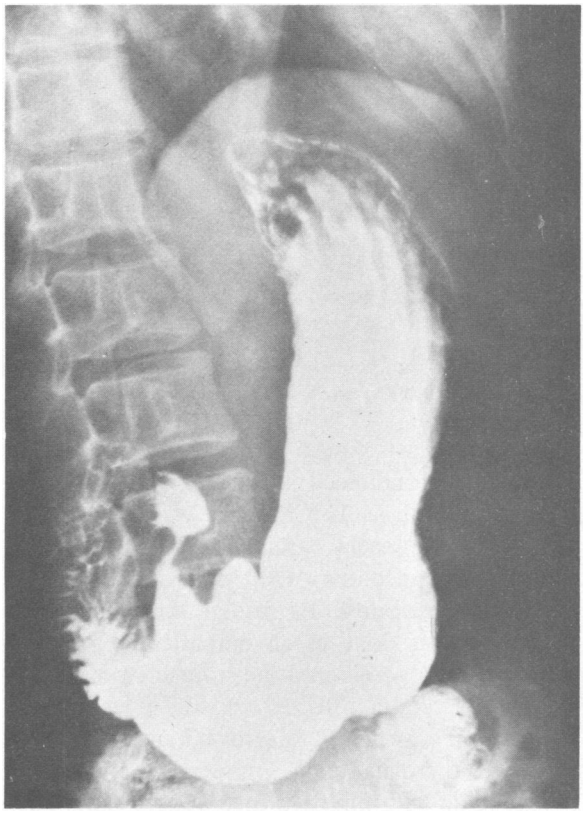

Figure 1.—Upper gastrointestinal series showing irregularity of the antrum and narrowing of the pylorus.

On physical examination, the patient was seen to be thin and appeared chronically ill. Aside from epigastric tenderness, the examination showed no abnormalities. Laboratory studies disclosed the following values: hematocrit, 38 percent; leukocyte count, 9.9×10^3; normal values for thyroxine (T-4), blood urea nitrogen, serum electrolytes, glucose and calcium; cholesterol, 152 mg per 100 ml; total protein, 6.6 grams per 100 ml, and albumin 3.8 grams per 100 ml. A Venereal Disease Research Laboratories test and an intermediate purified protein derivative were negative. No abnormalities were seen on an x-ray film of the chest.

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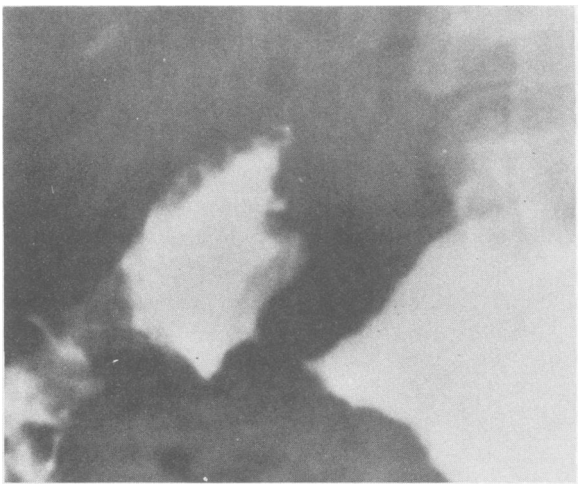

Figure 2.—Spot film from the patient's upper gastrointestinal series showing pronounced narrowing of the antrum and pylorus.

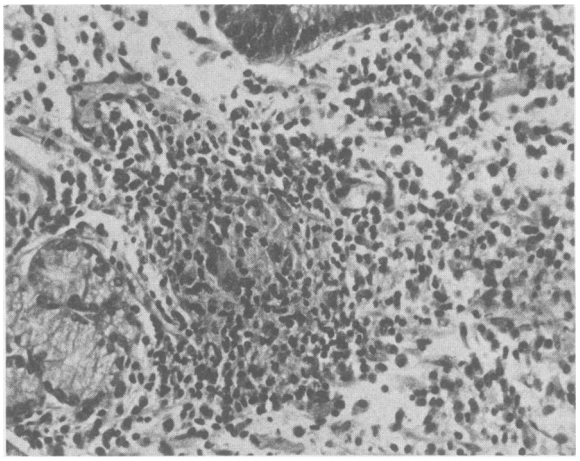

Figure 3.—Gastric biopsy specimen obtained by fiberoptic endoscopy showing antral mucosa and a focus of nonspecific granulomatous inflammation. (Hematoxylin and eosin; magnification, X240.)

An upper gastrointestinal series (Figures 1 and 2) showed pronounced antral deformity and decreased peristalsis. The findings were thought to most likely represent carcinoma of the stomach, with scarring secondary to peptic ulcer disease a less likely possibility. To resolve the uncertainty, endoscopy was done as an outpatient procedure. As the endoscope entered the stomach a moderate amount of retained secretions was encountered. The antrum was notably narrowed and contracted poorly. In the distal antrum, the mucosa was rigid and nodular with multiple ulcerations, some having a stellate appearance. The pylorus was narrowed and did not open sufficiently to allow the endoscope to be passed into the duodenum. Biopsy specimens and brushings for cytology were obtained.

Because of the presence of obstruction, the patient was admitted to the surgical service and treated with nasogastric suction. The preliminary reports of the pathology specimens were negative for tumor. At laparotomy the changes were consistent with peptic disease and a pyloric channel ulcer was described. A vagotomy and Finney pyloroplasty were carried out. Immediately postoperatively the patient did well and had no further problem with gastric retention. Later, examination of serial sections of the original biopsies (Figure 3) obtained at endoscopy showed foci of nonspecific, noncaseating granulomata with giant cells, consistent with Crohn's disease.

At first the patient did well and began to gain weight. However, she soon began to have problems with diarrhea, abdominal bloating, distention, lack of further weight gain, and foul smelling eructations. Upper gastrointestinal and small bowel series showed deformities of the pylorus and duodenum consistent with the surgical procedures carried out. The terminal ileum appeared to have irregular mucosa suggestive of inflammatory bowel disease. A barium enema study showed a normal colon with reflux into a normal terminal ileum. Because the patient's symptoms were thought to be due to active Crohn's disease, therapy was started with sulfasalazine (Azulfidine®), 500 mg three times a day, and prednisone, 30 mg every other day tapered to a maintenance dose of 20 mg every other day. Within one week of starting therapy, the diarrhea decreased and the other symptoms improved. Subsequently the patient has become asymptomatic. Repeat endoscopy after one year showed a postoperative stomach with grossly normal appearing mucosa. On random biopsy, only chronic inflammation was found. The patient continues to do well after 42 months with only periodic episodes of diarrhea.

Discussion

Granulomata have been found in a variety of diseases: sarcoidosis, tuberculosis, syphilis, cat-scratch fever, chronic ulcerative colitis, regional enteritis, berylliosis, silicosis, foreign body reactions, leprosy, brucellosis and histoplasmosis. In a review of the literature concerning granulomatous involvement of the stomach, several trends become evident. As in the original description of granulomatous involvement of the ileum, much debate and ascription to other diseases has oc-

curred. Fahimi[16] suggested that there were three different groups of patients. Group I comprises patients with documented granulomatous disease of the stomach with associated signs of sarcoid elsewhere. Group II is made up of patients with granulomas in the stomach and involvement of the bowel elsewhere which resembles regional enteritis. He also suggested a third group to be recognized as a distinct clinical entity, namely, those with isolated granulomatous gastritis in which there was gastric involvement only, with no signs of regional enteritis or sarcoidosis.

There have been many reports of gastric involvement by sarcoidosis.[16-19] If, as in the case reported by Orie,[17] there is pulmonary involvement and evidence of systemic disease, then it is reasonable to assume that granulomas encountered in the stomach are related to the sarcoid process. Likewise, the findings of granulomas in the stomach and involvement of bowel elsewhere warrants the diagnosis of Crohn's disease. The finding of isolated gastric granuloma without systemic disease makes the diagnosis of sarcoid or Crohn's disease less definite since the histology of the sarcoid granuloma is nonspecific.[20] Response to therapy cannot be used since both diseases behave similarly.

The first report that recognized granulomatous involvement of the stomach to be related to Crohn's disease was that by Ross.[5] Since then there have been more than 50 cases reported.[5-15] In considering the cases where sufficient data are available, there are several interesting aspects which clinically may help distinguish this involvement of the stomach by Crohn's disease.

Age and Sex

The youngest patient was 9 years old when diagnosis was made. The disease probably already had been present for some time. The oldest patient was 60 years old. The average age was 27 years. It should be noted that this is approximately the average age of onset of Crohn's disease elsewhere in the alimentary tract.[21] About two thirds of the patients reported were male.

Clinical

The two most common complaints in this group of patients were epigastric pain and weight loss. Nausea and vomiting were present in a large number, most often when obstruction was present. Diarrhea was present in about half of the patients. This feature occurred most often in those with involvement of other segments of bowel as well.

Radiologic

The most common findings on x-ray studies were decreased peristalsis, dilatation of the stomach and irregularity of the antrum. The radiologic diagnosis made most often was carcinoma of the stomach. Characteristic changes elsewhere in the bowel would suggest involvement of the stomach by Crohn's disease rather than carcinoma. In this review, in all but five of the cases there were findings of Crohn's disease elsewhere and in more than two thirds there was involvement of the duodenum.

Pathology

The pathologic findings are similar to those in Crohn's disease found elsewhere in the alimentary tract. In most cases biopsy specimens were obtained at surgical operation. The first report of a biopsy study showing a granuloma obtained through the biopsy channel of the fiberoptic endoscope was by Roseman.[9] Since then, there have been four additional cases.[10-12] The difficulty in obtaining biopsy specimens containing granuloma has been well pointed out by Danzi and coworkers.[11] However, the yield of granulomas can be increased by biopsy of microulcerations.[22] As in this case, examination of serial sections is also important.

Endoscopy

The findings at endoscopy were similar. These consisted of deformity of the antrum with narrowing, pale mucosa, cobblestone mucosa, mucosal inflammation and superficial ulceration. In addition, in some patients there were a nodular mucosa and polypoid masses with ulceration, either longitudinal fissures or stellate ulcerations.

Treatment

In most reported cases, surgical operation was done at some time in their course. Most operations were exploratory laparotomies or procedures for relief of gastric outlet obstruction. Course after operation was not predictable. Which is the best type of surgical procedure has not yet been resolved because of the limited number of cases and the difficulty in making the diagnosis preoperatively; the most popular surgical approach at present seems to be vagotomy and by-passing

gastroenterostomy.[23,24] The medical treatment of the disease is similar to that described for Crohn's disease in general.[25,26] The response to therapy with steroids is usually favorable and pyloric obstruction may be relieved without surgical intervention.

It seems that with a better understanding of the clinical signs and symptoms and a wider use of fiberoptic endoscopy and biopsy, more cases will be identified before operation and patients will be given a chance to improve as a result of steroid therapy alone.

Summary

In a patient thought to have carcinoma of the stomach on the basis of clinical and radiographic findings, fiberoptic endoscopy with biopsy was done for confirmation of the diagnosis. The patient was found to have Crohn's disease of the stomach. The average age of patients with this disorder is 27.3 years and two thirds are males. The presenting manifestations include epigastric pain, weight loss, nausea and vomiting. Radiologic findings are antral deformity and poor peristalsis of the stomach. Although in most patients with Crohn's disease of the stomach the disease also is present in other parts of the body, there may be isolated gastric involvement. Fiberoptic endoscopy with biopsy can establish the diagnosis of Crohn's disease of the stomach simply and safely.

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Lingual Thyroid in Two Members of a Family

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LINGUAL THYROID is a consequence of anomalous thyroid development. The frequency of this condition has been estimated to be approximately one in 4,000,[1,2] but the prevalence of lingual thyroid among asymptomatic euthyroid persons is not known. There is a high female to male ratio; in one series 87 percent of the cases were female.[3] According to one author,[4] about 15 percent of patients with this condition are hypothyroid. There has been one report of lingual thyroid in two siblings.[5] To our knowledge, no other instances of familial occurrence of this condition have been described. However, other types of developmental abnormalities of the thyroid have been noted among relatives of patients with lingual thyroid.[3,5]

This report describes the occurrence of lingual thyroid in a father and his son. The former was clearly hypothyroid by clinical and laboratory

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